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Remote site hemorrhage mechanism, management and outcome? Case report and literature review

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ABSTRACT

Background: Remote site hemorrhage (RSH) is a rare neurosurgical condition that occurs supra or infratentorial, away from the operation site after sudden decompression of high intracranial pressure (ICP) patients. Many surgeons have experienced such cases. Reviewing the literature shows that this kind of hemorrhage were mostly occurred in the cerebellum, has been named remote cerebellar hemorrhage (RCH).
Case description: Herein, we present a patient with remnant olfactory groove meningioma, in whom RSH was happened in the left temporal lobe just post-operation and subsequently RCH was occurred 48 h later. Finally, the patient was discharged without any neurologic deficit, after several days of conservative management.
Conclusion: Although no confirmed mechanism has been reported for RSH yet, excessive sudden cerebrospinal fluid drainage, whether intra-operative or post-operative, and consequently intracranial hypotension can be assumed as a major risk factor for it.

1. Background

Remote cerebellar hemorrhage (RCH) is a rare neurosurgical complication following supratentorial craniotomies, ranging between 0.08% and 0.6% according to the literature; however, there is no defined mechanism [1]. Considering two cases of RCH the senior surgeon (AT) previously has reported in his experiences, now we present another case with remote site Hemorrhage (RSH) on the left temporal lobe detected in just post-operation computed tomography (CT) and subsequently RCH, 48 h later. A RSH consists of an intracranial hemorrhage that occurs far from the operation site. Such a hemorrhage can occur in the supratentorial region (ST), infratentorial region (IT), or even in both regions simultaneously [2] like our case.

2. Case description

A 42-year-old man with complaints of headache, dizziness, and blurred vision for 10 years, and a history of brain tumor resection about one year ago in another center, presented with progressive mentioned symptoms during the last year showing no improvement. After imaging, he was scheduled for tumor resection due to remnant huge olfactory groove meningioma.

The patient was admitted to the neurosurgery department with complaints of headache, dizziness, and blurred vision. He didn't have any other history of medical disorders. In pre-operative neurological examination, the patient was conscious and his Glascow Coma Scale (GCS) was 15/15, pupils were normal in size and also reactive to light. The patient had complaints of decreasing left-sided visual acuity as well as also hyposmia which occurred after the previous craniotomy. The patient didn't have any medical disorder or abnormality in the laboratory data, including white blood cells (WBC), hemoglobin (Hb), platelet count, as well as coagulation profile.

In the latest brain magnetic resonance imaging (MRI), performed after the previous bifrontal craniotomy, in addition to post-operative left frontal encephalomalacia, an extra-axial midline mass in the anterior cranial fossa was demonstrated. The lesion was isointense in T1 and T2 images referring to peritumoral vasogenic edema. After contrast injection, gadolinium enhancement was depicted in the tumor (Fig. 1A–C).

The tumor was posteriorly located adjacent to the bilateral anterior cerebral arteries, causing compressed frontal horns of the lateral ventricles as well as the third ventricle due to mass effect. In just pre-operative spiral brain CT, a hyperdense patchy calcified mass was visible (Fig. 1D). The patient was scheduled for tumor resection in Jim supine position with head fixation via Mayfield. After bifrontal

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craniotomy on the previous surgical approach, the tumor was resected.

With no significant intraoperative complication or unusual event, gross total resection (GTR) lasted for nearly 140 min (skin to skin) and intraoperative blood loss was estimated 700 cc.

In just post-operation CT scan, in addition to multiple pneumocephalus, hyperdense patchy foci in the surgical field, supra- and infratentorial generalized edema, evidence of subarachnoid hemorrhage (SAH) in right Sylvian fissure (Fig. 2A), and nearly 5 cc cerebral hemorrhage in the left temporal lobe, named RSH, were noticed (Fig. 2B).

The patient was transferred to the Neurosurgical Intensive Care Unit (ICU) with GCS of about 12/15 (best eye movement score: 3, best motor score: 6, and best verbal score: 3) without any other deficit in the neurological examination.

In the first hours after surgery, vacuum drainage was changed to non-vacuum form due to CSF over drainage; the first post-operation Hb showed a decline of nearly 5 units, from 12.5 to 7.5, rose to 9.5 units after transfusing 1 unit packed cell.

On the second day post-operation, after CSF drainage control and also transfusing packed cell, the patient's GCS rose to 15/15, and second post-operation CT scan after 48 h showed decreasing generalized edema; however, cerebellar linear hemorrhage (zebra sign) was a new finding (Fig. 3A). According to decreasing CSF drainage, the non-vacuum drain was removed on the fourth day and control brain CT showed resolving cerebral and also cerebellar hemorrhage (Fig. 3B). Considering acceptable general condition and resolving process of hemorrhage in the next CT (Fig. 3C), the patient transferred to the neurosurgery department after 6 days and discharged from the hospital with good condition and also partially resolved Zebra sign on the 8th day control brain CT (Fig. 3D).

3. Discussion

RSH is defined as a hemorrhage distant from the operation site that has mostly occurred in supratentorial craniotomies and categorized in supra and infratentorial RSH. Chi wang et al. demonstrated that the incidence of IT RSH is more than ST, while the mortality rate of IT ones is higher [3]. While our case presented with synchronous IT and ST RSH, his outcome was good and didn't have any related complications after discharge.

RSH was reported in some neurosurgery centers as a complication of supratentorial operation and with less degree, after spinal surgery [4–7]. According to the literature, it seems that dural opening, intra- and/or post-operative CSF loss with consequent cerebellar sag, patient position during operation, preoperative aspirin usage, coagulation abnormalities, the intraoperative elevation of systolic blood pressure, and impaired venous drainage are proposed to have a causative role in RSH [4–6]. Among these, the most responsible cause of RSH proposed to be sudden CSF loss during or after the operation [4]. Coagulation abnormalities, hypertensive episodes, and patient position are less suspected mechanisms [4,5]. As mentioned above, our presented case didn't have any laboratory problem or drug usage before the operation, but losing large volume of CSF intraoperative and also after the operation, has strengthened this hypothesis that sudden decreasing in ICP has a causative role in RSH.

Although RSH is rare, in a patient with symptom deterioration after neurosurgical procedures, both on supratentorial or spine region, RSH should be considered [6]. It can be either a life-threatening complication or a simple one, probably according to the amount of hemorrhage [4].

In two previously reported cases in our center, a 57–year–old patient

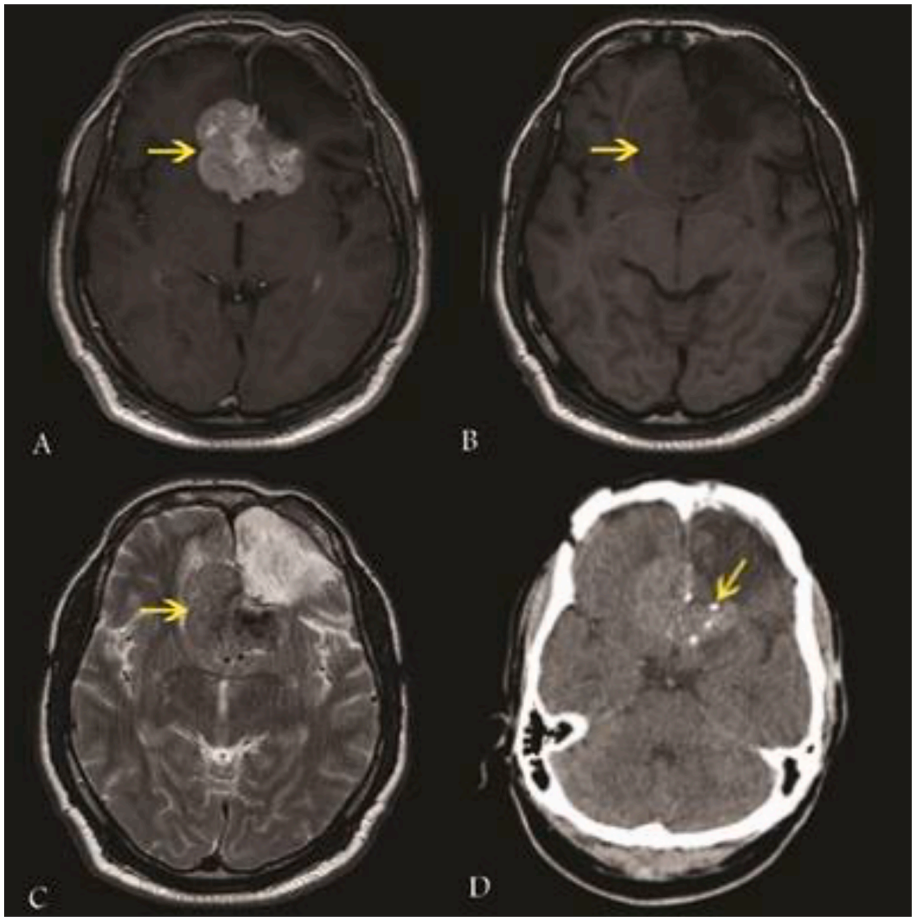

Fig. 1. Pre operation: A) Transaxial Gadolinium-enhanced MRI shows tumor remnant. B, C) T1 and T2 MRI images show an extra-axial mass in the anterior midline cranial fossa with peritumoral vasogenic edema. D) Transaxial view of the non-contrast CT. Hyperdense patchy calcified mass is shown by the arrow.

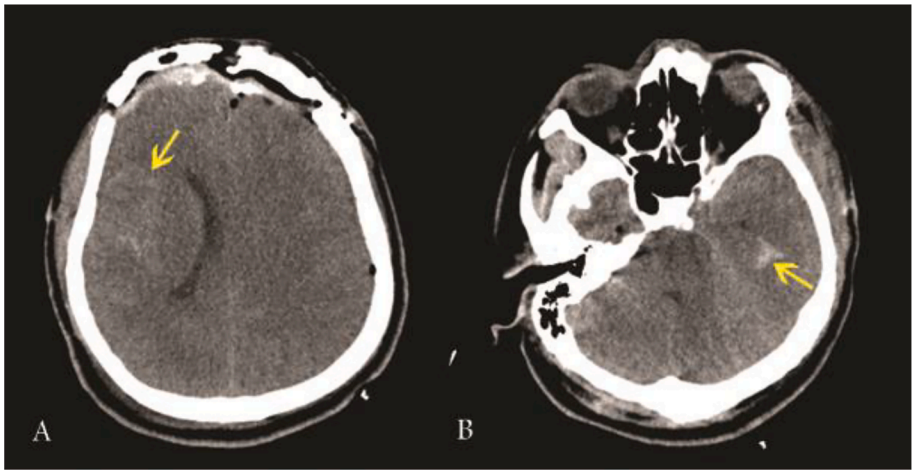

Fig. 2. First post-operation CT images. Transaxial CT views show right side Sylvian subarachnoid hemorrhage (A), as well as left temporal cerebral hemorrhage (B).

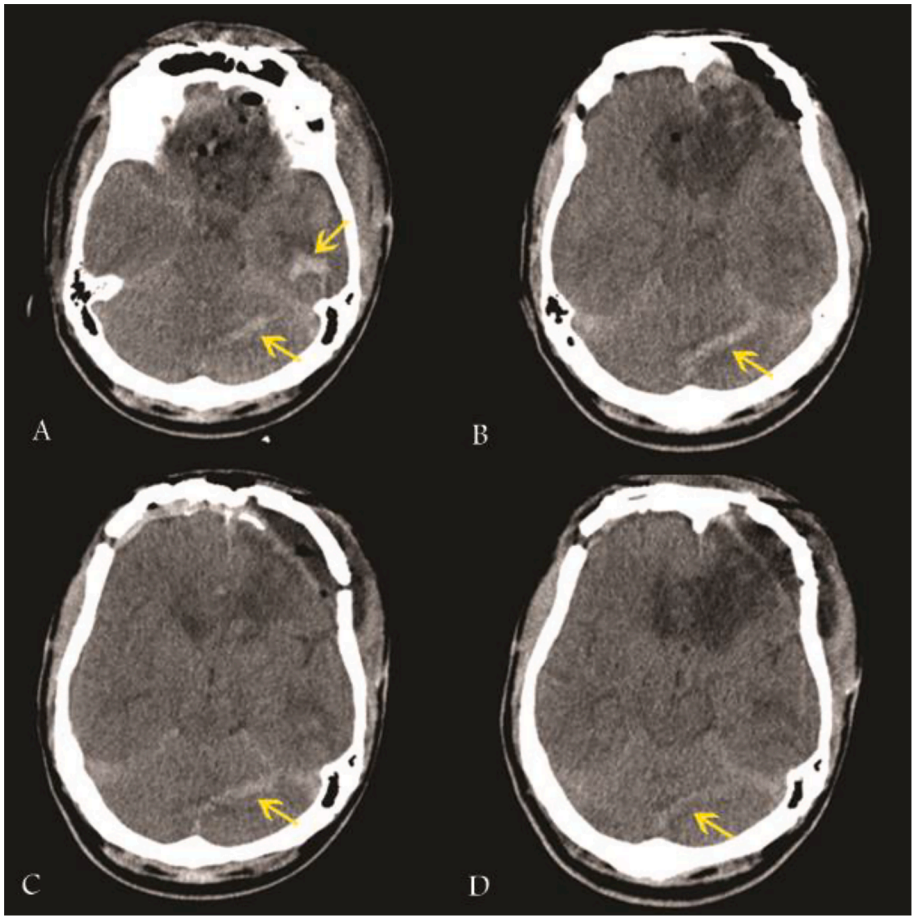

Fig. 3. Serial post-operation CT images showing Zebra sign resolving process. A) 48th hour CT shows left temporal and cerebellar linear hemorrhage (zebra sign). B, C) 4th and 6th days CT images show zebra sign resolving. D) 8th day CT image shows nearly resolved Zebra sign.

with sphenoidal wing meningioma demonstrated asymptomatic RCH 2 days after craniotomy. The other patient, a 21–year–old man with a huge intra-axial parietooccipital lesion, developed symptomatic RCH just after the surgery. The first patient had a good prognosis just as the present case, but the second one showed a poor prognosis [2]. In our previous cases, moderate CSF loss during the operation while cistern opening, normal blood pressure, platelets count, and coagulation profiles were noted. In the first patient, large post-operative CSF loss via vacuum was seen; however, in the second patient, RCH occurred just after the surgery. Just like the two mentioned cases, the recently reported patient had normal blood pressure, platelet count, and coagulation profiles.

Considering CSF over drainage in our three reported cases, and also other cases in the literature, avoiding CSF over drainage is recommended to prevent RSH in the high ICP patient who is defined according to pre-operation examinations and images.

According to the literature review, zebra sign in a patients post-operation CT scan, can indicate the probability of acute large CSF

leakage leading to cerebellum sag and cerebellar drainage vein occlusion or tear [3]. Brockmann et al. have presented a patient with Zebra sign in the brain CT scans with a corresponded blood clot within the subarachnoid space of the cerebellum at autopsy [8]. The striped pattern of zebra sign can be assumed a result from the pattern of subarachnoid bleeding due to the damaged cerebellar drainage vein, from which blood spreads to the cerebellar sulcus [3].

By reviewing the literature, ones can't find a defined protocol for managing the RSH. In the previous case reports, the surgeons have managed the RSH case by case, according to the mental status of the patient, as well as the volume and site of the bleeding [3]. As mentioned previously, we observed our patient via tight controlling the GCS, correcting hemodynamic instability, laboratory impairments, and also serial brain CT scans till resolving the RSH.

4. Conclusion

Increasing caution as well as recognizing the RSH lead the neurosurgeons to prevent this complication via restricted and slow CSF drainage by inserting lumbar drain just before opening the dura in high ICP patients according to pre-operation examination and images and also controlling post-operation CSF drainage via inserting non vacuumed drain.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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